

Neuroconstructivism to understand the effect of very preterm birth on language and literacy

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The preterm population is not characterised by an initial delay that recovers during development, but by atypical developmental trajectories, that can be understood through the neuroconstructivist approach. The neuropsychological profiles of very preterm infants show a great heterogeneity, depending on neonatal immaturity, medical complications, environmental, relational and social factors. The developmental trajectory of very preterm children is described in relation to the acquisition and consolidation of language and literacy from early infancy to adolescence. Indexes for early individuation of language and literacy delays, as well as for planning focused interventions, are discussed.

Keywords: demands, moderately preterm, word learning mechanism

Rethinking preterm birth with the neuroconstructivist approach

Preterm birth is defined by the World Health Organization when babies are born alive before 37 weeks of gestational age (GA); the rate of preterm birth ranges from 5% to 18% across 184 countries (March of Dimes, 2012). Different sub-categories of preterm birth were defined in function of GA (extremely preterm, <28 weeks; very preterm, from 28 to <32 weeks; moderate to late preterm from 32 to <37 weeks, March of Dimes, 2012).

Preterm birth constitutes a risk condition for cerebral, physical and neuropsychological development (March of Dimes, 2012) that can be understood through the neuroconstructivist framework (Sansavini, Guarini & Caselli, 2011). Neuroconstructivism argued that development is determined by the interplay of multiple biological and environmental constraints interacting at different levels (genes, brain, body, and environment, Karmiloff-Smith, 1998; Westermann et al., 2007). In preterm infants atypical constraints occur in a critical period of rapid

development of the neural system (Volpe, 2009) determining an atypical developmental trajectory (Sansavini, Guarini, & Caselli, 2011) resulting from the adaptation to multiple altered constraints (Karmiloff-Smith, 1998). In other words, preterm birth forces brain and body to adapt to an artificial environment characterised by hypo-stimulation (e.g., absence of rhythmic and kinaesthetic stimulations) and over-stimulation (e.g., light, noise, painful and invasive medical interventions, Sansavini, Guarini, & Caselli, 2011) affecting cerebral, physical and neuropsychological development. Neuroconstructivism can also explain the wide heterogeneity observed in the neuropsychological profiles of preterm infants (Guarini & Sansavini, 2010; Sansavini, Guarini, & Caselli, 2011) in function of different levels of neonatal immaturity, medical complications and the environmental and social characteristics to which the child is exposed, such as the level of parental education and their responsiveness to the child's communicative signals (Bozzette, 2007).

This risk condition associated to preterm birth can affect the development of communication and language, since the auditory system and the neural circuits involved in the development of language are particularly vulnerable in the last trimester of gestation, when preterm newborns live in an extra-uterine artificial environment (Vohr, 2014). The early exposure to an artificial environment of a biologically immature system increases the risk of atypical development pathways (Sansavini, Guarini, & Caselli, 2011). This vulnerability can be amplified by environmental characteristics (e.g., exposure to a noisy environment and lack of rhythmic stimulation, such as maternal heart rate) and fewer communication opportunities with parents and people who take care of the newborn (Sansavini et al., 2017; Vohr, 2014).

The atypical developmental trajectory of very preterm children in communication, language and literacy is described in the following paragraphs of the present chapter. In the last paragraph, focused interventions proposed to preterm children and their parents are described in order to indicate good practices for creating an enriched environment and enhancing protective factors.

Effects of preterm birth on communication and language development

As shown by many studies conducted in several countries and by recent meta-analyses and reviews, preterm birth has an impact on the development of communication and language since early childhood and persists until adolescence (Barre et al., 2011; Sansavini, Guarini, & Caselli, 2011; Sansavini & Faldella, 2013; Sansavini et al., 2017; van Noort van der Spek et al., 2012). Most effects have been found on extremely preterm and very preterm infants, but some also on moderate (32–33 weeks GA) and late preterm (34–36 weeks GA) infants. Language

difficulties of very preterm children are often associated with early perceptual, cognitive, communicative, and motor problems that may have cascading effects on later more complex abilities (Sansavini, Guarini & Caselli, 2011).

Speech perception skills, such as discrimination, recognition and tuning on the phonetic units and the prosodic characteristics of the mother tongue, as well as word segmentation, develop in the first year of life (Bosch, 2011). When corrected age has been used (i.e., referring to the expected date of birth of 40 weeks GA), only slight delays have been found in very preterm infants, showing that the development of speech perception skills is much affected by neurobiological maturation (Bosch, 2011; Peña et al., 2010).

By contrast, early gestural and vocal production is less developed between the first and second year of life in extremely preterm and very preterm children with respect to full-term children, even when using corrected age (Benassi et al., 2016; Sansavini et al., 2011, Sansavini, Bello et al., 2015). Gesture and word production at 12 and 18 months is predictive of word production at 24 months indicating the importance of monitoring and supporting these skills from early stages of development (Sansavini et al., 2011; Stolt et al., 2014).

The ability to share attention with the caregiver on a common focus is fragile in extremely preterm and very preterm children and strongly affected by mother-infant interaction modalities. Synchrony, symmetric co-regulation and cooperation in mother-infant interaction, constitute important protective factors for communication development (Forcada-Guex et al., 2006; Landry et al., 1996; Sansavini, Zavagli et al., 2015). In particular, maternal contingent and highly relevant responses (i.e., those with a repeated label) following infants' spontaneous communicative behaviors support communication development in extremely preterm infants, being related with their receptive and expressive communication skills at 12 months and expressive communication skills at 24 months (Benassi et al., 2018).

Fine and gross motor skills are also often compromised in very preterm and extremely preterm children since the early years of life, affecting the ability to explore objects that is crucial to the construction of cognitive and linguistic categories (Ruff et al., 1984; Sansavini et al., 2014; Zuccarini et al., 2016). Recently, studies on extremely preterm infants have found concurrent and longitudinal relationships between fine motor skills and communication development: global fine motor skills were positively associated with communication skills, specifically with pointing and representational gestures, at 12 months (Benassi et al., 2016), whereas active object exploration behaviors at 6 months were associated with gesture and language abilities at 12 months (Zuccarini et al., 2018) and with linguistic and cognitive skills at 24 months (Zuccarini et al., 2017).

Studies conducted in different countries with extremely preterm and very preterm children between the second and third year of life, have shown a less

advanced development with respect to full-term children in phonology, lexicon and grammar, both in comprehension and production (D'Odorico et al., 2011; Foster-Cohen et al., 2007; Sansavini et al., 2010, 2011, Sansavini, Bello et al. 2015; Stolt et al., 2009, 2017). The risk of language delay in very preterm children increases from 20–25% at two and a half years to 30–35% at three and a half years, whereas in the same period it decreases from 13% to 7% in full-term children (Sansavini et al., 2010), highlighting the importance of monitoring language development of preterm children during this period. Language delay in very preterm children is often associated with delays in basic information processing skills, such as lexical processing speed (Marchman et al., 2016) and in cognitive skills, such as executive functions (Aarnoudse-Moens et al., 2009; Dall'Oglio et al., 2010), with a higher severity in case of neurological damage (Woodward et al., 2009). These findings suggest the importance of assessing basic general abilities alongside specific language skills to outline the neuropsychological profile of these children and schedule early interventions in multiple developmental domains.

Lexicon, grammar and phonological skills continue to be affected by a very preterm birth during preschool and school age (Guarini et al., 2009, 2010, 2016) with scores ranging from 0.38 to 0.77 standard deviations (SD) lower than those of full-term children (Barre et al., 2011; van Noort-van der Spek et al., 2012). Towards the end of preschool age, difficulties also arise in phonological awareness (Guarini et al., 2009) and pragmatic skills, for example in the narrative of figurative stories (Guarini et al., 2016). These difficulties are often associated with cognitive difficulties, particularly with working memory skills (Guarini et al., 2016; Sansavini et al., 2007; Wolke & Meyer, 1999).

The differences between very preterm and full-term children in the above mentioned language skills often remain up to adolescence even in the absence of severe neurological impairment or disability and regardless of parental socio-economic level. However, as far as lexical comprehension is concerned, a study showed a recovery in adolescence, associated with high level of maternal education, presence of both parents in the family, and absence of neurosensory damage, suggesting that protective environmental factors may have compensatory effects on some aspects of linguistic development (Luu et al., 2011; van Noort-van der Spek et al., 2012).

Some delays have also been found in moderate and late preterm children: their language skills, even if better than those of very preterm children, were lower than those of full-term children (Putnick et al., 2016). Since individual differences in language development in the preterm population become gradually stable between 2 and 4 years of age and remain stable up to 8 years, it is important to detect early language delays and plan targeted interventions starting from preschool age (Putnick et al., 2016).

Acquisition and consolidation of literacy

Several reviews revealed that preterm birth affects not only language but also literacy skills. Delays were found in the acquisition and consolidation of reading and spelling processes with different developmental trajectories in function of gestational age (Guarini & Sansavini, 2010; Sansavini, Guarini & Caselli, 2011). In extremely preterm children delays emerge in primary school and up to secondary school, with a mean of 15 points lower than that of their full-term peers (Johnson et al., 2009). A different picture emerges in very preterm children who have greater difficulties in the spelling process (-0.76 SD), than in the reading process (-0.48 SD; Aarnoudse-Moens et al., 2009; Guarini et al., 2010). Delays in spelling are already present in the acquisition phase, with lower performances in handwriting due to difficulties in fine-motor coordination and visual-motor integration (Feder et al., 2005), and they persist up to secondary school (Chaudhari et al., 2004). Some difficulties were also described in moderate preterm children (de Jong et al., 2012), even if a partial recovery in reading decoding (Rose et al., 2011) and spelling (Tideman, 2000) has been observed in secondary school.

However, most studies have been conducted with native English-speaking children, exposed to an opaque orthography. Very few studies have been carried out in preterm children exposed to a language with a transparent orthography, such as Italian, highlighting the importance of interpreting the results obtained according to the specificity of the mother tongue. Italian very preterm children showed delays in spelling, with errors in word, non-word and sentence tasks at 8 years of age. In reading, speed was affected whereas decoding was accurate as that of full-term peers (Guarini et al., 2010). These results suggest that the effect of preterm birth on reading in a language with transparent orthography is particularly evident in speed, as already found in dyslexic children native speakers of orthographically regular languages (Zoccolotti et al., 1999).

As revealed by a recent meta-analysis, delays in very preterm children involved not only decoding, but also comprehension of written texts (Kovachy et al., 2015), revealing that difficulties in reading comprehension were not recovered, but they increased with increasing age-related difficulties (Kovachy et al., 2015).

Starting from these considerations, the process of reading and spelling of preterm infants required to be monitored both in the acquisition phase and in the consolidation phase, since a higher rate of impairments across multiple curriculum areas are frequent among preterm children (Litt et al., 2005; Pritchard et al., 2009). In addition, the role of cognitive variables, such as processing speed and executive functions, should be taken into account, since they mediate the role of preterm birth in learning difficulties (Loe et al., 2012; Rose et al., 2011). In addition, extremely preterm children are at high risk for comorbidity between learning

disabilities and intellectual disabilities (Johnson et al., 2016). The role of preterm birth on literacy was also mediated by language acquisition, since phonological awareness and grammar comprehension showed significant correlations with literacy processes at 8 years of age (Guarini et al., 2010). In addition, phonological awareness and lexicon, before entering primary school, are predictive of literacy skills two years later (Guarini & Sansavini, 2012). Eventually, additional variables may influence learning, such as motivation, and self-efficacy, since preterm children appear to benefit less from learning opportunities and show more motivational problems (Taylor et al., 2000).

Interventions

The studies described in the previous paragraphs outline the importance of monitoring the development of oral and written language of preterm infants and, in particular, of very preterm and extremely preterm infants, from early infancy to adolescence. Follow-up programs should be implemented in order to detect developmental delays and to propose customized interventions from the first years of life onwards (Sansavini & Faldella, 2013).

As shown by a recent review (Spittle et al., 2015), early intervention programs begin within the first 12 months of life and are focused on infant development and/or parent-infant relationship. Notwithstanding the great heterogeneity of these interventions, their positive impact on later cognitive outcomes until at least preschool age has been shown (Spittle et al., 2015). Indeed, preterm infants involved in these programs reported higher cognitive scores than infants who received a standard follow-up program (0.32 SD in infancy and 0.43 SD at preschool age). However, these differences were no longer evident at school age and adulthood (Spittle et al., 2015). Interestingly, interventions focused on both infant development and parent-infant relationship show a higher positive impact on later outcomes (Spittle et al., 2015).

NIDCAP (Newborn Individualized Developmental Care Assessment Programme) is an early intervention programme widely used in Neonatal Intensive Care Units (NICU). This is a family-centered and personalised intervention aiming at enhancing parental care and preterm infant well-being and reducing stress conditions of the NICU (Als, 2009; Als et al., 2012). Significant positive effects of NIDCAP on neurodevelopmental outcomes of preterm infants were found between 9 and 12 months (Jacobs et al., 2002), whereas evidence at later ages is still scarce (Ohlsson & Jacobs, 2013).

Benefits on infants' self-regulation, improving sleep-wake cycle, arousal modulation and sustained exploration of the environment at 6 months, have also

been shown through the implementation of Kangaroo Mother Care, which aims at promoting skin-to-skin contact between preterm infants and their caregivers (Feldman et al., 2002). A significant positive effect of this early intervention has been found both in increasing maternal attachment behaviour and child autonomic functioning in the first year of life and on cognition, executive functions and mother-child reciprocity up to 10 years (Feldman et al., 2014). The Kangaroo Mother Care was endorsed by the World Health Organization in 2003 as an intervention highly recommended and as an effective solution to reduce deaths among preterm infants (March of Dimes, 2012).

Early intervention programs focused on enhancing parent-infant relationship and parent coping have shown positive effects on infants' neurodevelopmental outcomes at least up to 36 months of age (Vanderveen et al., 2009). A study by Brooks-Gunn et al. (1992) showed that an intervention programme on parent-child relationship improved cognitive (visual-motor and spatial skills) and language skills of low-birth weight preterm infants at 24 and 36 months, with some differences in treatment efficacy in function of birth-weight and ethnic group. Parent-infant interventions have shown positive effects also on parental outcomes, reducing maternal anxiety and depressive symptoms and increasing their self-efficacy (Benzies et al., 2013).

In recent years, interventions to improve specific competences of very preterm children have been designed.

For instance, infant massage, proposed in the NICU following a specific protocol, accelerates brain activity and visual function, such as visual acuity, in preterm infants (Guzzetta et al., 2009, 2011).

Recent research suggests that encouraging parents to talk and sing to their newborn during hospitalization in the NICU is very promising for language development (Filippa et al., 2013). Very preterm infants increase vocal production between 32 and 36 weeks when parents talk to them and the amount of parental language in NICU is associated with child language and cognitive development at 7 and 18 months (Caskey et al., 2014) suggesting that a positive early auditory experience contributes to cerebral maturation and neuropsychological development.

Some interventions were also conducted to improve specific cognitive functions that are particularly vulnerable in pre-school preterm infants, such as working memory. Very preterm children, who followed a computerized cognitive training at pre-school age, improved several skills that had an impact on later achievement, such as auditory attention, phonological awareness, visual and verbal memory and sentences repetition (Grunewaldt et al., 2013).

Further research is needed to replicate these studies and examine the long-term effects of interventions on developmental trajectories of preterm children.

Conclusions

The present chapter describes preterm birth and its effects on language and literacy development within the neuroconstructivist framework. Since preterm birth exposes the newborn to atypical constraints, an atypical developmental trajectory is shaped with possible delays in language and literacy from the first years of life to adolescence. Some abilities may be particularly affected with a high variability in function of neonatal immaturity, medical complications and social risk factors. An important role of speed processing and executive functions is also described, revealing strict relationships among domains, as already described in other populations with atypical development (Karmiloff-Smith, 1998). As suggested by the neuroconstructivist framework, positive body experiences, adequate environmental stimuli, and positive social interactions can improve the outcomes of preterm infants, indicating the importance to design and promote effective early intervention.

Authors contributions

Conceived and designed the content of the manuscript, wrote and revised the paper: AG AS. Wrote the paragraph on interventions: MZ AG AS.

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